Surgical resection of a large recurrent pelvic arteriovenous malformation using deep hypothermic circulatory arrest

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Transcatheter embolization has emerged as the treatment of choice for pelvic arteriovenous malformations (AVMs), because surgical resection may be difficult and is associated with a high recurrence rate. We report a patient with a large recurrent pelvic AVM in whom transcatheter embolization was not feasible. This patient underwent surgical resection of the AVM, which was accomplished with deep hypothermic circulatory arrest. Early postoperative angiography demonstrated a small amount of residual AVM, which was successfully embolized with microcoils. Follow-up magnetic resonance angiography at 2 months showed no residual AVM. In cases where surgical resection of an extensive AVM is required, deep hypothermic circulatory arrest offers the distinct advantages of performing the resection in a bloodless field and enabling adequate visualization of important adjacent structures. (J Vasc Surg 2004;39:1348-50.)

Pelvic arteriovenous malformations (AVMs) are rare congenital lesions characterized by a large number of arterial feeding branches.[1] Histologically, they are composed of abnormal dysplastic vessels.[1] Surgical resection is often difficult, with the potential for exsanguinating hemorrhage and damage to surrounding structures.[2,3] Recurrence of the AVM is common with incomplete resection. Ligation of large feeding vessels is to be averted, because it makes subsequent embolization more difficult, and ligation of these vessels is never curative.[1,2] Accordingly, transcatheter embolization has become the treatment of choice.[2] Preoperative embolization of AVMs has also been used as an adjunct to decrease intraoperative blood loss.

We describe a patient in whom surgical resection of a large recurrent pelvic AVM was deemed the only option available, because of innumerable feeding vessels originating from multiple intraabdominal vessels and from the femoral vessels. Because of concern about excessive hemorrhage and the need for extensive dissection in the pelvis, we elected to perform the operation with deep hypothermic circulatory arrest.

CASE REPORT

This 43-year-old woman was referred in April 2003 after her gynecologist noted an increase in the size of a pelvic AVM. In 1998 an asymptomatic pelvic AVM had been found at physical examination. An arteriogram demonstrated feeding vessels arising from both the right and left internal iliac arteries. The AVM was treated with ligation of a branch of the right internal iliac artery. A hysterectomy was performed to assist in exposure.

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In 2000 a pulsatile mass was palpated along the right lateral wall of the vagina by a gynecologist. No radiographic studies were performed.

In March 2003 the patient was referred to us when the gynecologist noted that the pelvic mass had increased in size. A thrill and bruit were present. The patient had no symptoms.

A magnetic resonance angiogram (MRA) demonstrated a large pelvic AVM, with effacement of the right posterolateral wall of the bladder (Fig 1). An arteriogram demonstrated "innumerable" branches arising from both internal iliac arteries, the right L4-L5 lumbar arteries, the middle sacral artery, the rectosigmoid branch of the inferior mesenteric artery, and a branch from the right common femoral artery. The venous phase of the arteriogram demonstrated huge pelvic veins. Our interventional radiologists believed that transcatheter embolization was not feasible in this patient, because of the large size of the pelvic AVM and the innumerable feeding vessels.

The patient had no symptoms from the AVM, neither pelvic symptoms, such as pain, dysuria, urinary frequency, or dyspareunia, nor any cardiovascular manifestations. Because we thought the AVM should be treated, we did not perform any cardiac studies. However, physical examination did not demonstrate any signs of right-sided heart failure. No cardiomegaly was noted on a chest x-ray.

Surgical resection was deemed advisable, even though the AVM was asymptomatic, because of the growth of the AVM. Because of the risk of significant hemorrhage and the need for extensive dissection in the pelvis, we thought this procedure would be best accomplished with deep hypothermic circulatory arrest.

At operation, bilateral ureteral stents were placed to aid in identification of the ureters. The previous lower midline abdominal incision was reopened to just above the umbilicus. We were able to resect a significant portion of the AVM before initiating cardiopulmonary bypass. There was no encapsulation of the AVM. At this point we had spent approximately 5 hours on the resection. The remaining portion of the AVM was adherent to the sigmoid colon and bladder. We believed the remaining resection should be accomplished with deep hypothermic circulatory arrest.

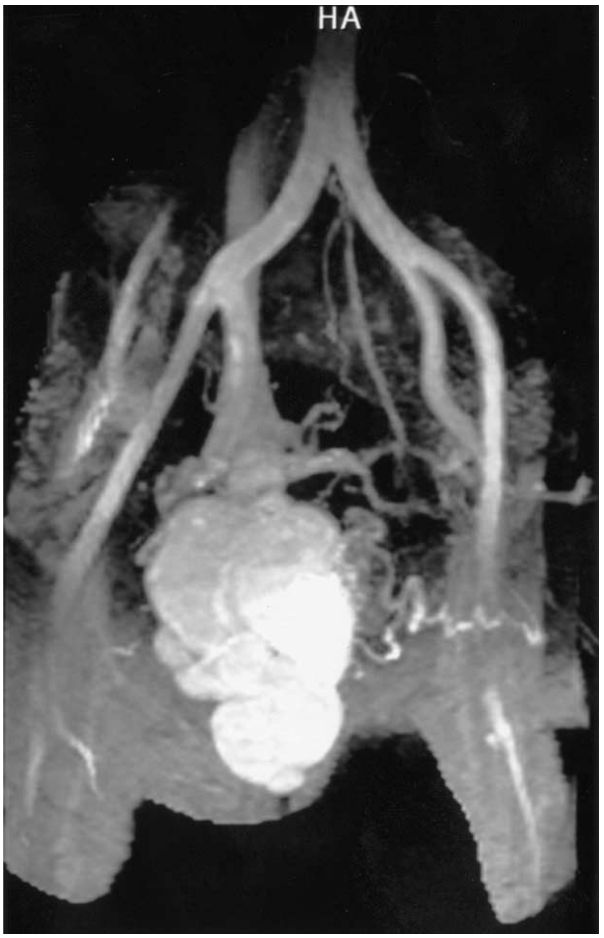

Fig 1. Magnetic resonance angiogram demonstrates a large pelvic arteriovenous malformation, with effacement of the right posterolateral wall of the bladder.

Through the same midline abdominal incision, the infrarenal aorta and inferior vena cava were exposed. Heparinization was then accomplished with 3 mg/kg of heparin. Purse-string sutures were placed just proximal to the aortic and inferior vena cava bifurcations for cannulation. A long 33/30F Carpentier bicaval cannula (Medtronic, Minneapolis, Minn) was passed up the inferior vena cava into the right atrium by means of the Seldinger technique. Similarly, through the infrarenal aortic purse-string, a 20F Fem-Flex cannula (Edwards Life Science, Irvine, Calif) was passed into the infrarenal abdominal aorta. Cardiopulmonary bypass was then instituted, and the arterial blood was immediately cooled to 15°C. After 30 minutes of cooling, the tympanic temperature was reduced to 17°C, and the rectal temperature to 16°C. Just before cessation of cardiopulmonary bypass, 40 mEq of potassium was administered as a bolus into the pump circuit to obtain cardiac standstill. In a completely bloodless field, the remainder of the large AVM was resected. The period of circulatory arrest was 10 minutes.

Cardiopulmonary bypass was then reinstituted at 2 L/min of flow, and the blood was gradually rewarmed to 22°C. Both common iliac arteries were temporarily occluded during this phase of rewarming, as the remaining bleeding sites were ligated. After controlling most bleeding sites, the blood was then continually rewarmed to 37°C, and cardiopulmonary bypass was increased to 4.5 L/min, which is essentially normal cardiac output for this patient's body size. During the rewarming phase at 33°C, one additional large arterial branch was identified that was bleeding, and the pump flow was again discontinued for 3 minutes to ligate this vessel. Cardiopulmonary bypass was resumed at 4.5 L/min, and rewarming was completed. Cardiopulmonary bypass was discontinued, and protamine was given. Total pump time was 145 minutes, of which 13 minutes involved circulatory arrest. No platelets or other coagulation factors were used.

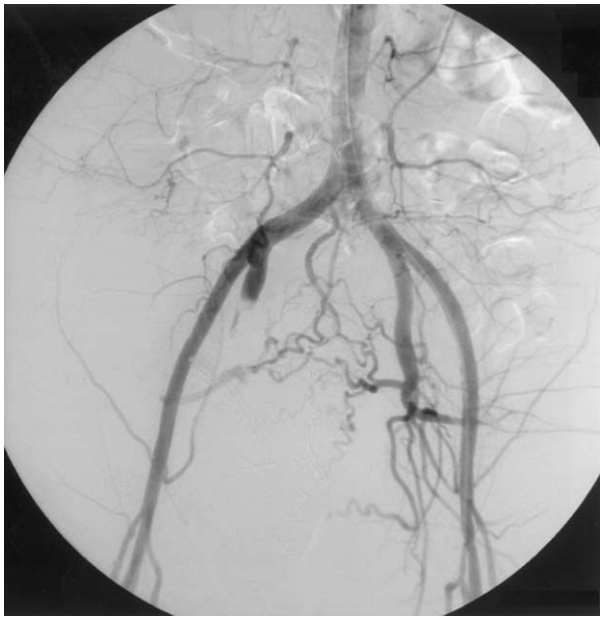

Fig 2. Angiogram obtained on postoperative day 3 demonstrates a small amount of residual arteriovenous malformation in the pelvis, which was embolized with microcoils.

At this point there was some oozing in the pelvis, but no significant bleeding. A closed suction drain was placed in the pelvis, and was brought out through a "stab" wound in the right lower quadrant. The patient received 4 units of blood during the operation. The operation lasted 8 hours 42 minutes.

Postoperatively, the patient did well. An angiogram obtained on postoperative day 3 demonstrated a small amount of residual AVM in the pelvis, which was embolized with microcoils (Fig 2). At 2-month follow-up an MRA demonstrated no residual AVM. We plan to obtain an MRA at 6 months, and then yearly.

DISCUSSION

Pelvic AVMs may manifest with symptoms of pain, hemorrhage, hematuria, dyspareunia, or congestive heart failure, or symptoms may develop because of compression of adjacent pelvic structures.[2] Small asymptomatic AVMs that do not increase in size may be safely observed.[4] We elected to treat the AVM in our patient, even though it was asymptomatic, because it had grown and was compressing the bladder wall.

Transcatheter embolization has emerged as the preferred treatment for pelvic AVM.[1,2] Multiple embolizations may be required. Embolization has also been used as an adjunct before surgical resection, to decrease intraoperative bleeding. Our interventional radiologists have had extensive experience with embolization in the treatment of AVMs and uterine fibroids. After consultation with the interventional radiologists, we believed this AVM should be treated operatively with deep hypothermic circulatory arrest. In addition, embolization is not without complications. Jacobowitz et al[2] reported intractable hematuria after embolization, thought to be secondary to bladder ischemia.

Attempts at surgical excision may be complicated by significant hemorrhage, which also increases the risks for damage to adjacent tissues. Flye et al[3] reported two patients who died after an attempt at surgical resection of an AVM.

Two previous reports have described the use of deep hypothermic circulatory arrest to treat an AVM in the mandible and a recurrent posttraumatic arteriovenous fistula in the pelvis.[5,6] Both patients had undergone previous unsuccessful attempts at surgical resection. With deep hypothermic circulatory arrest, the pump can be turned off, enabling the resection to proceed in a bloodless field. The pump can be turned on briefly, as in our case, to identify any residual bleeding and enabling a more complete resection. In our experience with deep hypothermic circulatory arrest in repairing arch aneurysms, coagulopathy has seldom been encountered. The other distinct advantage of deep hypothermic circulatory arrest is that it enables the surgeon to identify adjacent structures, thereby minimizing the risk for damage to those structures.

Some investigators have recommended maintaining low flow (500 ml/min) with profound hypothermia, because this may increase the duration of safe ischemia while not producing bleeding at the operative site.[5] We have not found this to be necessary. The risk for neurologic injury with deep hypothermic circulatory arrest is low, provided the period of circulatory arrest is less than 1 hour.[7,8]

There are no firm indications as to when deep hypothermic circulatory arrest should be used in the treatment of AVM. We agree with other authors who advocate transcatheter embolization as the primary treatment method.[1,2] However, there are cases in which experienced interventional radiologists may believe that embolization may not be feasible, or cases in which embolization has been associated with complications and further embolization is deemed hazardous.[2] Patients with significant right-sided heart failure may benefit from surgical resection with deep hypothermic circulatory arrest, which would provide more prompt resolution of heart failure than multiple embolizations.

CONCLUSIONS

Deep hypothermic circulatory arrest is a valuable adjunct in the treatment of large AVMs in which surgical resection is necessary. Resection of the AVM in a bloodless field minimizes the risks for damage to adjacent vital structures, and may enable more complete resection. Periods of circulatory arrest up to 60 minutes are well tolerated.

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